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Oral mucosal involvement in visceral leishmaniasis

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ABSTRACT

Leishmaniasis affects both the visceral and cutaneous tissues in body. Oral Mucosal involvement in leishmaniasis is rare and is often overlooked. We present a case 17 year old boy from the north east region of Bihar who has a history of visceral leishmaniasis one year back, came to the department of oral surgery for treatment of persistent oral ulcers. Oral examination did not give any diagnostic information while systemic examination revealed enlarged spleen and low grade fever. Patient was screened for leishmaniasis by rK39 based immunochromatographic strip test which came to be positive. Biopsy of the ulcer as well as splenic and bone marrow aspirate confirmed the presence of leishmaniasis. Patient was administered Amphotericin B for 20 days following which significant clinical and haematological improvement followed.

1. Introduction

Leishmaniasis affects 12 million people worldwide and is seen in nearly 88 countries. Incidence of leishmaniasis is highest in India, Bangladesh, Nepal, Brazil and Sudan. The oral mucosal involvement is rare in leishmaniasis. Mucosal leishmaniasis (ML) affects the upper respiratory tract and/or oral mucosa leading to mucosal ulcerations mainly in the hard or soft palate[1]. There are only few reported cases of mucosal involvement in leishmaniasis[2], in some cases it was seen in post leishmaniasis treatment[3]. This condition is often misdiagnosed by health care professionals as being fungal infections or oral cancer. A high suspicion of oral leishmaniasis should always be kept in patients presenting with persistent gingival ulcers or hypertrophy, non-responsive to therapy, especially those coming from an endemic zone[4]. Early diagnosis and institution of anti-leishmanial therapy can prevent an irretrievable damage to oral and nasal tissues in such cases[5].

2. Case report

A 17 year old male reported to the department of dental surgery complaining of persistent, multiple, painful ulcers on the base of tongue and on the palate since 30 d, non-responsive to any treatment. The ulcers were painful and bled on touch, causing a difficulty in eating. He came from an area endemic for kala-azar and there was a past history of treatment for the same one year back. Patient denied any use of tobacco or alcohol, extramarital sexual relation or intravenous drug abuse. He looked pale and emaciated. Oral examination revealed multiple small ulcers on the base of the tongue (Figure 1) and palatal mucosa, with marked erythema. These were nodular, swollen, raised with irregular margins and all were smaller than a centimetre in size. Teeth and gingiva were in good health. Neck examination revealed bilaterally enlarged, non-tender submandibular lymph nodes. Systemic examination revealed fever of 101.4 Fahrenheit and palpable spleen. Haematological investigations revealed anemia (Hb 7.6 g/dL), thrombocytopenia (platelet count 70 000/mL), slight leukocytosis (total leukocyte count 12 000/mL) with polymorphic predominance and albumin:globulin (A:G) ratio being 2.6:5.0. Renal function tests and urine microscopy was normal. VDRL and HIV tests were negative. Residence in an endemic area, past history of treatment for kala-azar, palpable splenomegaly and evidence of anemia and A:G reversal prompted us to go for rK-39 immunochromatographic strip test. Not to our surprise, it was positive for kala-azar. Aspirates from spleen and bone marrow (Figure 2) also depicted the presence of Leishmania-Donovan (LD) bodies, confirming the diagnosis. Scrapings from the oral ulcer also revealed the presence of amastigotes on histopathological examination. Patient

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was given *iv*. Amphotericin B (1 mg/kg, qod) for 20 d. During treatment no complication was seen. From the 3 rd day of the start of the treatment oral mucosal lesions begin to resolve and disappeared completely by the 8th day. Fever charting of the patient showed slight rise in fever on the 4th and 5th day followed by rapid decline to the normal level.

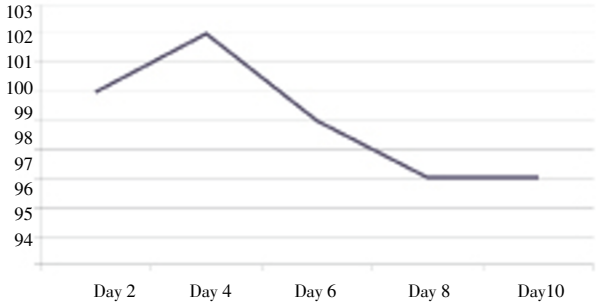

Figure 1. Depicting the temperature charting of the patient during initial phase of amphotericin B therapy.

Patient was discharged on the 22nd day from the start of the treatment. General blood picture was normal at the time of discharge and splenomegaly had almost regressed. Patient was followed up monthly for three months and three monthly for one year. He remained afebrile, with no evidence of mucosal lesions or organomegaly.

3. Discussion

Visceral leishmaniasis(VL) is caused by protozoa lieshmania. Around 20 species of leishmania has been described. Three major forms are visceral, cutaneous and mucocutaneous, each of which is attributed to different species of leishmania. In the most parts of the world the Phlebotomus species is responsible for transmission, while in USA Lutzomyia species is the responsible species of sandfly. 500 000 new case of VL are diagnosed every year. 90% of which is seen in five countries. These are Bangladesh, India, Brazil, Sudan and Nepal. In India, Bihar, adjoining areas of West Bengal, Jharkhand and Uttar Pradesh account for half of the total cases of leishmaniasis throughout the world. ML can be a primary disease or could follow an episode of VL. The two have been speculated to be connected as they are caused by the same parasite. On the contrary, ML in Sudan has been attributed to a different subspecies of the *Leishmania donovani (L. donovani)* complex and majority of cases in that region presented with isolated mucosal form not accompanied by the visceral form[6-8].

Earlier reports of visceral leishmaniasis presenting as oral ulcers in are present[9-12]. In our case the patient had previous history of leishmaniasis. This was confirmed by his previous bone marrow aspiration report from the district hospital where he was admitted, but he did not have complete record of treatment given during hospitalization. Persistent infection due to inadequate treatment is not uncommon. Patients ignore complete medical treatment either due to shortage of money or feeling of well being following initial therapy. However, possibility of re-infection could not be ruled out in this case. We chose amphotericin B for therapy as the cure rates are excellent for patients from Bihar state[13].

A high suspicion of oral leishmaniasis should always be kept in patients presenting with persistent oral ulcers non-responsive to therapy, especially those coming from an endemic zone and having a past history of leishmaniasis or persistent fever and splenomegaly. Appropriate laboratory tests and histopathological examination is of utmost importance for ruling out other diseases clinically mimicking ML, such as oronasal tumors, leprosy, histoplasmosis, and aspergillosis. Early diagnosis and institution of anti-leishmanial therapy can prevent an irretrievable damage to oral and nasal tissues in such cases.

Conflict of interest statement

We declare that we have no conflict of interest.

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